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Original Article

AMELOBLASTIC FIRBO-ODONTOMA ASSOCIATED WITH CALCIFYING ODONTOGENIC CYST – A CASE REPORT & REVIEW OF THE LITERATURE

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ABSTRACT

Hybrid odontogenic tumours are rare and present with numerous histopathological patterns. Odontogenic tumours such as ameloblastoma, ameloblastic fibroma (AF), ameloblastic fibro-odontoma (AFO), and odontoma are uncommonly found in conjunction with odontogenic cysts such as calcifying odontogenic cyst (COC), glandular odontogenic cyst (GOC), and odontogenic keratocyst. The case of a 19-year-old female patient with an expansile, bony hard swelling connected with the left posterior maxilla is presented. It was showing mixed radiopaque and radioluent nature. Histologically, on incisional biopsy, it was found to be an ameloblastic fibroodontoma. After complete excision of the lesion, along with an ameloblastic fibroodontoma it was also associated with a cystic fluid filled cavity which on microscopic examination was found to be a Calcifying odontogenic Cyst. We believe it is important to report COC with AFO with full clinical and pathological evidence because there are few examples of COC with AFO cited in the English literature.

KEYWORDS: Ameloblastic Fibro-Odontoma, Calcifying Odontogenic Cyst, Hybrid Tumors & Odontogenic Tumors

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INTRODUCTION

Hooker developed the term ameloblastic fibro-odontoma (AFO) to describe an uncommon benign slow-growing expansile epithelial odontogenic tumour with odontogenic mesenchyme, which he discovered in 1967. [1] AFO is defined by the World Health Organization (WHO) as a neoplasm constituted of proliferating odontogenic epithelium embedded in a cellular ectomesenchymal tissue that mimics dental papilla, with varying degrees of inductive change and the creation of dental hard tissue..[2] AFO incidence is rare and maximum is less than 4.6 % in children.[3] Radiographic findings resemble complex odontoma as tiny teeth surrounded by radiolucent capsule.[4]AFO is usually treated by enucleation and prognosis is excellent. [2]

In 1962, Gorlin et al. classified calcifying odontogenic cyst as a separate entity. [5] There are several subtypes, each with its own set of clinical and histological characteristics, as well as biological behaviour. COC

usually manifests as an intraosseous lesion, however, it can also manifest as an extraosseous lesion. COCs are mostly cystic and non-neoplastic, although they can also show as solid lesions, and some of them are cancerous. In 2005, the World Health Organization termed it Calcifying Cystic Odontogenic Tumor (CCOT).

Radiographs may demonstrate a well-defined, uni or multi-locular radiolucent lesion associated with unerupted teeth that may or may not include different amounts of radiopaque material. COC resembles complex or compound odontoma, ameloblastoma, odontoameloblastoma, ameloblastic fibroma, ameloblastic fibro-odontoma, and the adenomatoid odontogenic tumour histologically. [6]

This article is about a case of an unusual occurrence of AFO associated with the COC in the left maxillary posterior region.

CASE REPORT

For 15 days, a 19-year-old female patient has complained of swelling and discomfort in the upper left jaw region. Throughout the procedure, the swelling remained painless. The swelling started out little, around the size of a betel nut, but it became larger over time. The patient stated that chewing was difficult, but that no intraoral discharge had occurred. There was no major medical or familial history for the patient.

Extraoral examination indicated a bulge with diffuse borders in the left maxillary area. It was approximately extended supero-inferiorly from a horizontal line passing through the floor of the orbit to a horizontal line joining the corner of mouth and the pinna of the ear. The skin overlying the swelling was smooth, non adherent to underlying swelling, & no change in color.

The swelling was bony hard on palpation and slightly elevated temperature from the surrounding tissues. No regional lymph nodes were palpated.

Swelling ran from the distal surface of the left maxillary first Premolar to the tuberosity intraorally. It was discovered that the buccal and lingual cortical plates had expanded. The expansion was concentrated on the buccal side, completely obliterating the buccal vestibule.

On probing, the swelling was bone firm and non-tender. Hard tissue examination revealed that 26 was displaced buccally and that 27 and 28 were absent. There was no movement with any of the teeth.

In the left maxillary alveolar region, an orthopantomograph (OPG) and CT scan revealed a radiolucent lesion with some flecks radiopacity extending mediolaterally from the distal surface of the maxillary first premolar to the distal surface of the maxillary tuberosity and superoinferiorly from the floor of the orbit to the occlusal surface of the maxillary ridge. Impacted 28 is also observed which is embedded in the tumor mass. In one of the CBCT view, the left maxillary sinus was obliterated due to the tumor mass.

To rule out the fibro osseous lesions, the serum levels of alkaline phosphatase and acid phosphatase were evaluated which were within normal limits.

On incisional biopsy, we received five tissue specimens which were varying in dimension, with grayish white in color and firm in consistency somewhere the gritty hard consistency is noted.

The incisional biopsy revealed lesional tissue consisting of odontogenic epithelial cells grouped in the form of nests, islands, and tennis racquet shaped islands on histopathological inspection. With polarity reversal, the peripheral cells are cuboidal to low columnar in form. The centre cells resemble those of a stellate reticulum, and some are spindle-shaped. These tumour cells have a hyalinization zone around them. The stroma of the connective tissue is loose and fibromyxoid, similar to the primitive stroma of dense papillae. Only a few extravasated RBCs are visible. At times, bony trabeculae might be detected on the periphery.

Decalcified section showed lesional connective tissue irregular arrangement. The dentin at places is in the form of tubular dentin seen in cross and longitudinal sections. Odontogenic epithelial islands are seen in the central part of the dentin matrix. Huge enamel spaces are seen throughout. At places, enamel prism like structures are seen. The peripheral portion showed loosely arranged fibromyxoid stroma.

Based on these cilicopathological correlations, the patient is diagnosed as a case of AMELOBLASTIC FIBRO ODONTOMA.

The patient is then operated under general anesthesia.

After nasoendotracheal intubation and induction of general anesthesia, skin over the operation side is painted with betadine and spirit. Intraoral gingival crevicular incision with releasing incision in the left maxillary first premolar to third molar region is given. Flap is raised, tumor is completely curetted completely along with the extraction of impacted third molar and enucleation of lining & tumor is done. The cavity is cauterized chemically. Flap is reconstructed & sutured with 3-0 vicryl suture material. Extra oral pressure dressing is done.

After excision of the tumor, we received 7 large bits of the tissues. The shape of these bits was irregular. The largest bit was 4.5 cm X 4 cm in size, the smallest was 1.5 cm X 15 cm in size. Rests of the bits were intermediate size. These bits were whitish gray in color. The consistency of these bits was firm and gritty hard at some places.

During grossing of the one specimen, we encountered a cystic cavity, containing a liquid material of a brownish colour. Few calcified hard spicules are noted on the inner side of the cystic cavity.

The histopathological examination of these specimens revealed similar findings as that of incisional biopsy.

Additionally, histopathological examination revealed a cystic cavity lined by stratified epithelium having 2-3 cell layers in thickness. It also shows the well defined eosinophilic and enlarged, ballooned, ovoid epithelial cells. The connective tissue capsule is composed of bundles of collagen fibers and fibroblast. A few odontogenic rest cells are also noted in the capsules.

The final diagnosis of ameloblastic fibroodontoma with calcifying odontogenic cyst was reported.

DISCUSSIONS

Ameloblastic fibroma and similar lesions are neoplasms made up of proliferating odontogenic epithelium embedded in a cellular ectomesenchymal tissue that mimics dental papilla, according to the World Health Organization. Ameloblastic Fibroma (AF), ameloblastic fibrodentinoma (AFD), and ameloblastic fibro-odontoma are among the mixed odontogenic tumours that include these abnormalities (AFO). 8-11 the odontomas, which are non-neoplastic malformations comprising fully calcified or mineralized dental tissues, are included in this group of mixed odontogenic tumours. 9,11. It's difficult to tell if these mixed odontogenic tumours are neoplasms or stages of a non-neoplastic developmental abnormality because of

the evident problems in distinguishing them from developing odontoma.

We present a case of ameloblastic fibro odontoma with a minor cystic component that had some characteristics of a COC.

Initially, Praetorius (1975) mentioned a link between the COC and the AFO. Then, in 1977, Allan G. Farman et al. described a case of coc linked with the AFO that was produced secondary to the cyst formation. 12

A comparable example of ameloblastic fibro-odontoma was reported by Matsuzaka K et al (2001), in which AFO is assumed to have developed from the lining epithelium of COC and is noteworthy for its high tendency toward infiltration.[13]

Constantino L M et al (2008) also mentioned a similar association in their review of mixed odontogenic tumours. [14]

A hybrid odontogenic tumour with traits comparable to ameloblastic fibro-odontoma, calcifying odontogenic cyst, and adenomatoid odontogenic tumour was described by Philips MD et al (2010). [15]

Because of the obvious difficulties in understanding the similarities and differences in the nature and development of these tumours, it's difficult to say whether these calcifying odontogenic cysts have features of odontogenic tumours that developed secondary to the cysts, or whether the cysts were secondary to pre-existing odontogenic tumours. Takeda et al. (1990)16 were sure that the cysts arose spontaneously, but Praetorius et al. (1981)17 believed that the tumour developed from the cyst wall. They proposed that the calcifying odontogenic cyst arose from decreased enamel epithelium or remains of odontogenic epithelium in the follicle, gingival tissue, or bone and that it was a unicystic process**.**

HirshbergA et al (1994)[18] reported several possibilities regarding the pathogenesis of COC and Odontoma which develop from the odontogenic epithelium independently or in association with each other.

CONCLUSIONS

Hybrid odontogenic neoplasms present a high degree of variance in histological patterns and under reported worldwide. Most of these tumours can be surgically enucleated with the least chance of recurrence unless there is any malignancy associated. Long term follow up should be emphasized.

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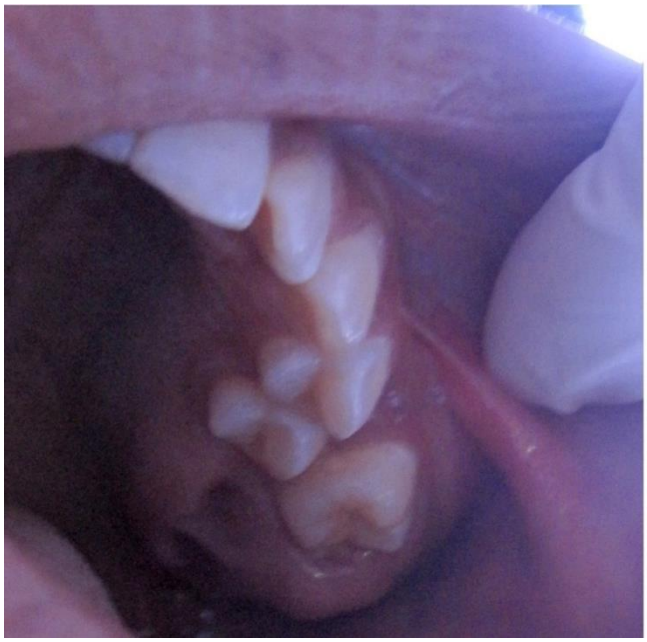
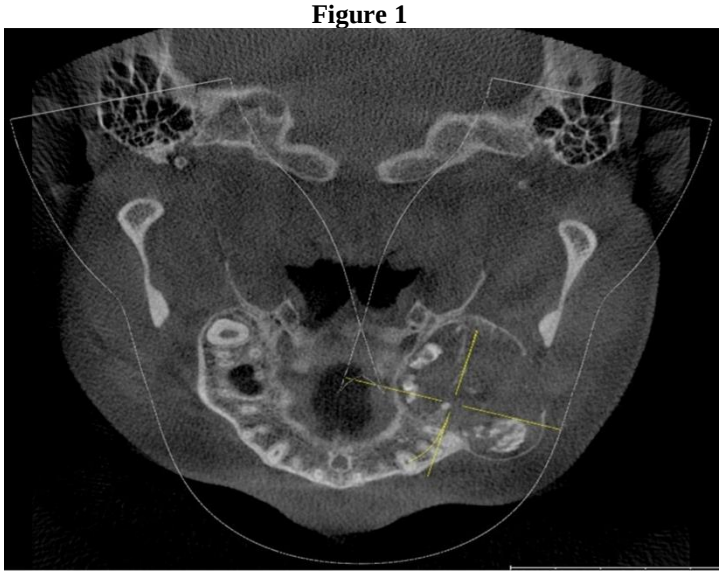


Figure 1



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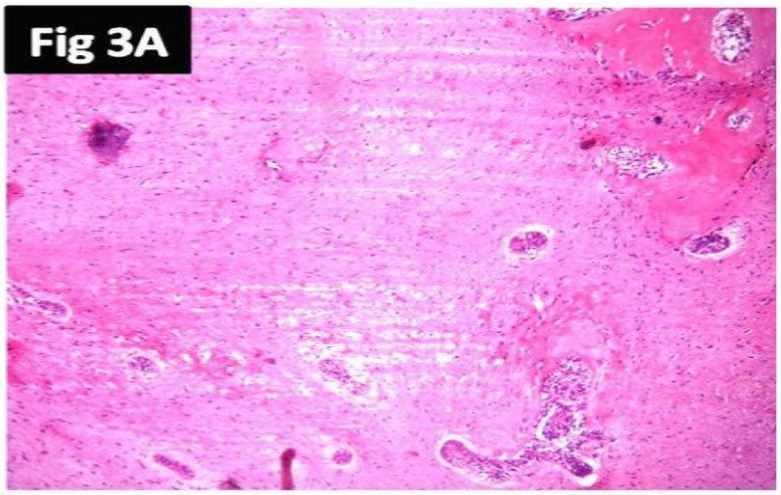
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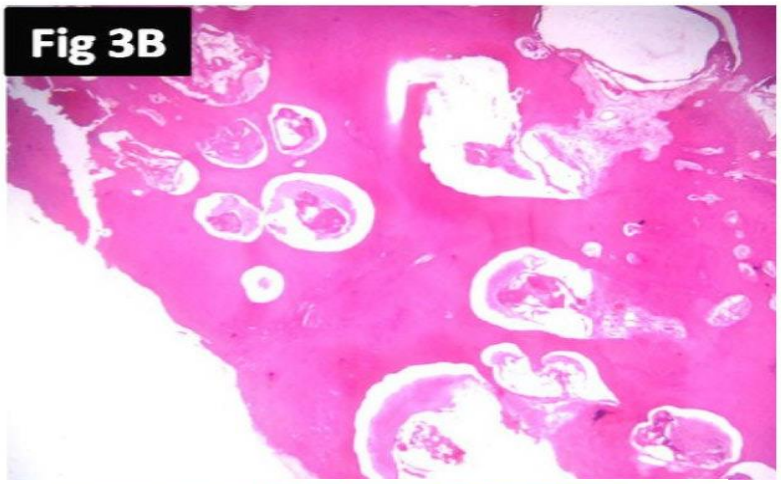
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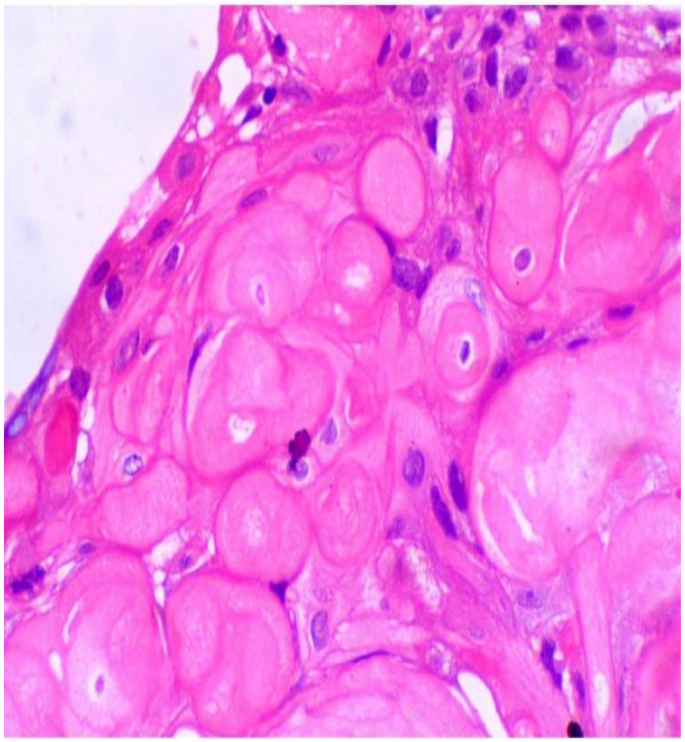

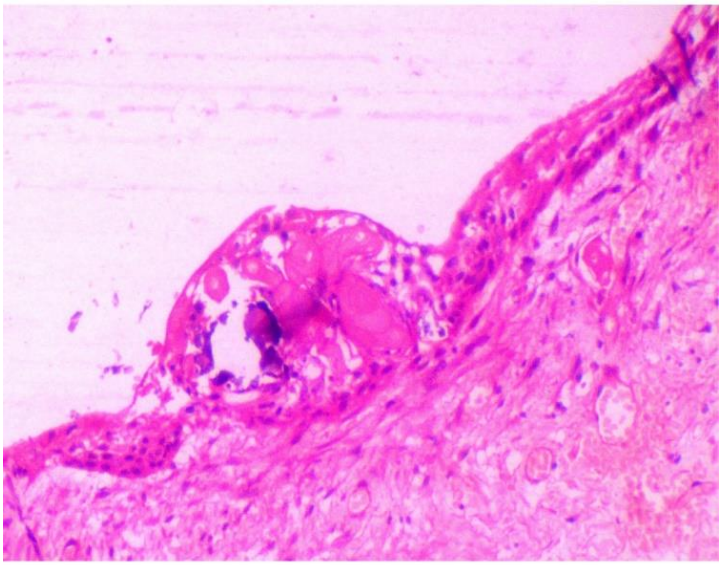

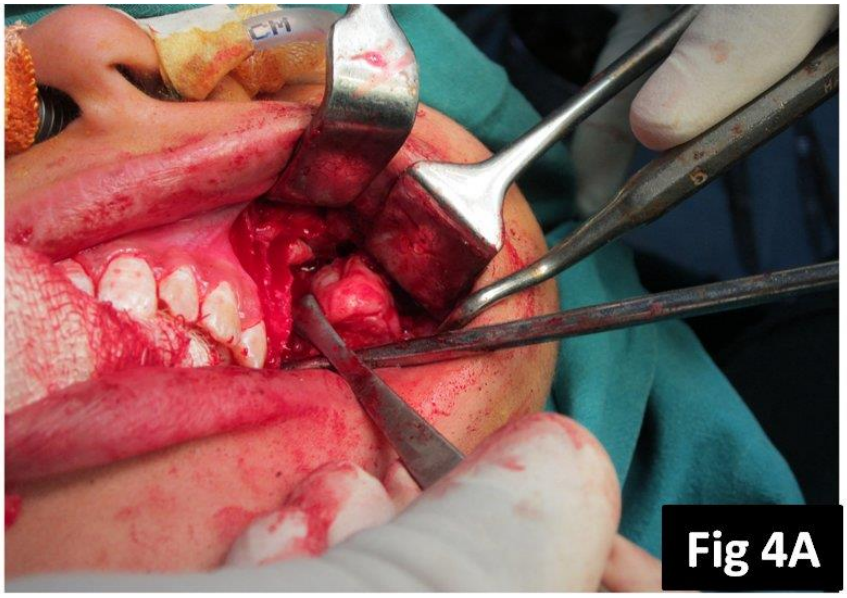

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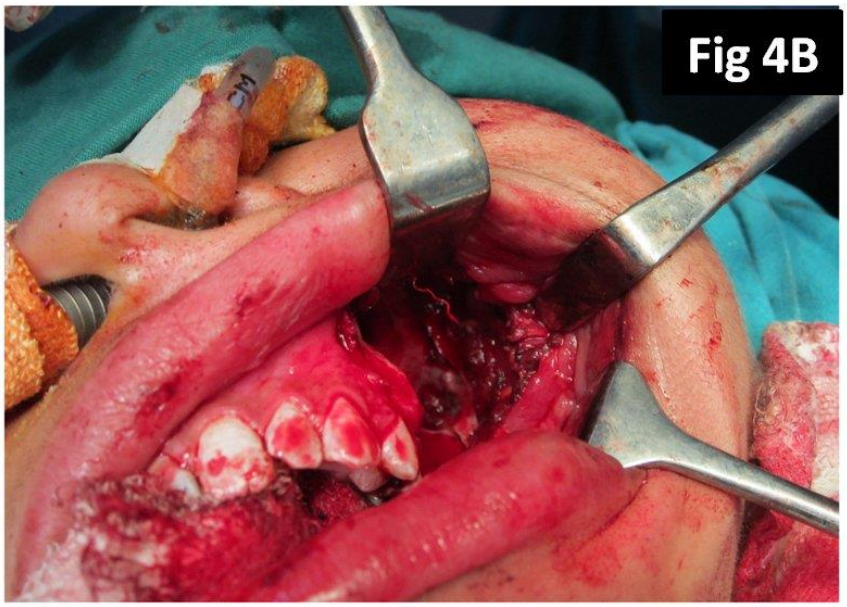
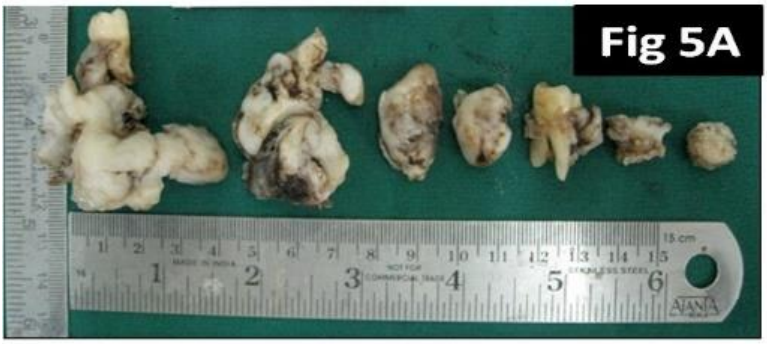


Fig 4B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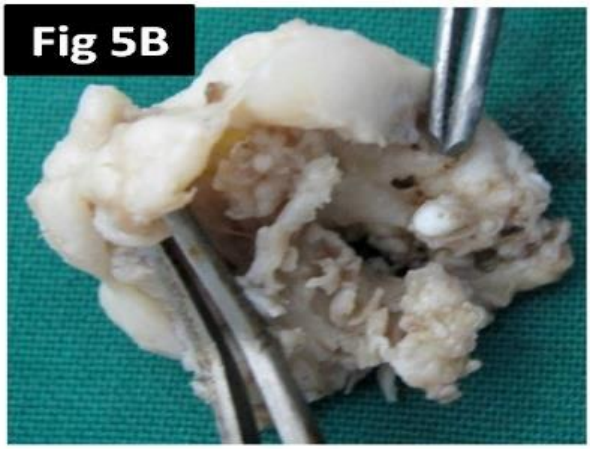
Fig 5B